

# The History of Aphallia and the Intersexual Challenge to Sex/Gender

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A great deal of media attention has recently been focused on the treatment of intersexuality (a variety of anatomical and physiological conditions historically called hermaphroditism). The tragic story of David Reimer (known as the “John/Joan case”) first captured public attention in 1997 and was followed by numerous television documentaries on intersex conditions.<sup>1</sup> Jeffrey Eugenides’ novel *Middlesex* featured an intersex heroine/hero and won a Pulitzer prize.<sup>2</sup> The Intersex Society of North America (ISNA), founded in 1993 (before Reimer’s case was publicized), has also done much to publicize the treatment of intersex conditions. The media has focused on dramatic cases of sex reassignment, like that of David Reimer who, in fact, was not born with an intersex condition. ISNA, on the other hand, has struggled to refocus public and professional attention on the more common issue of genital “corrective” surgery rather than neonatal sex reassignment, which is rarely necessary or recommended by doctors. The media has also tended to present intersex as a single phenomenon or associate it with transsexualism when, in fact, there are a variety of syndromes of genital atypicality and the vast majority of affected individuals have no indeterminacy about their gender identity. ISNA has struggled to convince the public that intersex is not an elective identity position but a variety of objective, biological conditions. Along these lines, a group of ISNA affiliated doctors and board members have advocated a significant nomenclature change: to discontinue terms employing “hermaphrodite” in favor of the term “disorders of sex development” (DSD).<sup>3</sup> This change is also being endorsed by an international group of medical experts in the field.<sup>4</sup> The use of DSD would make clear that “intersex” states are medical conditions affecting the development of the sex organs, not matters of gender identity, dual sex, or sex reassignment. DSD is a brand new term, even to the medical profession; therefore, I will continue to use (and tease apart) the term “intersex” in the remainder of this essay.

The frequency of “intersex” births is uncertain, partly because there is no standardized registry of congenital disorders, but also because there is no medical consensus on what conditions to include under the rubric of intersex or what degree of anatomical atypicality constitutes an intersex state. For example, hypospadias (malformation of the male urinary outlet) is fairly common (1 in 150–300 live male births), but severe hypospadias with female appearance of the genitals is extremely rare.<sup>5</sup> So,

## The History of Aphallia and the Intersexual Challenge to Sex/Gender

is hypospadias, in general, an intersex condition or not? Whether one argues for broad or narrow inclusion criteria has as much to do with politics as anatomy. Using broad inclusion criteria, the prevalence of any form of atypical genitalia may be as high as 2 percent of live births. More conservatively, extreme genital ambiguity for which surgery is considered occurs in 0.1–0.2 percent of births.<sup>6</sup> For purposes of comparison, the adult incidence of diabetes in the US in 2004 was 0.7 percent. Diabetes is a widely known disorder that attracts much media attention and research funding.<sup>7</sup> Therefore, depending on how one calculates the incidence of intersex conditions, they may affect an even greater number of people than diabetes. Whatever the numbers, intersex conditions can cause great physical and emotional distress, yet have been largely ignored by the general public until recently.

The delivery of a child with ambiguous or atypical genitalia poses issues that have long challenged surgeons, endocrinologists, and geneticists. There is far less literature by mental health professionals on the topic, no doubt because the secrecy about intersex interventions keeps patients ignorant of their condition or ashamed to discuss it. ISNA and other intersex support groups have done much in the past decade to lift the veil of secrecy and shame around intersex issues. People with androgen insensitivity syndrome (AIS), congenital adrenal hyperplasia (CAH), hypospadias, and other intersex conditions formed their own support groups in the 1990s and finally have been able to share their emotional and medical experiences.<sup>8</sup> These groups have also encouraged doctors to be more open in discussing intersex issues with parents and affected children to help them cope with the psychological and medical challenges they face.

The inspiration for this essay is a little boy who has been my psychotherapy patient since the age of ten. He was born with an anatomical anomaly called penile agenesis: the failure to form a penis in early embryonic development. The mainstream thinking in the 1980s was that boys with “micropenis” (less than 2.5 cm stretched length in neonates) or aphallia had inadequate genitals for male functioning. Therefore, doctors recommended to my patient’s mother that the infant be assigned female sex, undergo surgical feminization, and be raised as a girl. Shortly after birth his scrotum was shaped to resemble labia and the undescended testes were removed. Eighteen months later surgeons removed the small phallus. His mother named the infant Maria. However, Maria rebelled against feminine clothes and toys from an early age, and his mother finally let him take on a boy’s name and gender role at age five. Ever since then Mario has been an iatrogenic transsexual: a child with male gender identity in a surgically feminized body. Mario’s family and developmental history are quite complex. I have described his history and treatment in great detail in more appropriate clinical contexts.<sup>9</sup> In this essay, I am less interested in the details of his specific case than in trying to make historical sense of his predicament and how this history broadly affects infants with intersex conditions. I first review the history of hermaphroditism in order to trace the evolution of medical theorizing and treatment of intersex conditions. The history of penile agenesis, in particular, is closely tied to dramatic changes in the conceptualization of gender identity.

## The History of Genital Ambiguity: From “Hermaphroditism” to “Intersexuality”

For two millennia, ambiguous genital anatomy has been referred to under the term “hermaphroditism.” Since classical times, physicians and biologists have been fascinated by hermaphroditism as it was seen not only as a curiosity of nature but as a phenomenon central to the understanding of animal generation, embryological development, and sexual differentiation. Biological theories of hermaphroditism also reflect broader historical and cultural constructions of sex and gender. In trying to survey the medical history of hermaphroditism we can identify five major approaches to sexing the body based on:

- 1 External genital anatomy (from classical texts until the nineteenth century) determined by:
  - (a) the balance of sexual essences (Hippocrates, Galen)
  - (b) opposed sexual essences (Aristotle)
- 2 Gendered behavior including sexual orientation (eighteenth to nineteenth century)
- 3 Gonadal histology (nineteenth century)
- 4 Genetics (twentieth century)
- 5 “Optimal gender” (1950s).

These approaches are not historically discrete – diverse elements of these general perspectives are sometimes incorporated in any particular theory of intersexes and their treatment. For example, adult gender identity and behavior continue to be a factor in deliberations about the best sex assignment of intersexed neonates within the “optimal gender” treatment paradigm. Nevertheless, it is useful to distinguish these conceptual divisions.

The term *hermaphrodite* refers to the mythic character of Hermaphroditos. In Book IV of the *Metamorphoses*, Ovid described Hermaphroditos as a male embodying the ideal masculine qualities of his father, Hermes, and the feminine qualities of his mother, Aphrodite. In Ovid’s telling, the nymph Salmacis spied Hermaphroditos bathing and immediately fell madly in love. She begged the gods to unite them forever. Wielding their cruel sense of humor, the gods took Salmacis’ request literally and melded her body to that of Hermaphroditos.

It is not clear whether this mythical hermaphroditic body possessed both male and female parts or was of an intermediary sex. This question has been central to biological and philosophical debates about hermaphroditism ever since antiquity.<sup>10</sup> Hippocratic texts (fourth century BC) explained that hermaphroditism represented an intermediate form in the spectrum between the pure female and the pure male.<sup>11</sup> The Aristotelian tradition, which held that male and female were of fundamentally opposite natures, insisted that hermaphrodites possessed a supplemental sexual appendage opposed to their fundamental sex, analogous to a sixth digit or a third nipple.

From the classical period onward, hermaphroditism was critical to teratology – literally, the study of so-called monstrosities, marvels, and prodigies of nature. The modern era of experimental teratology began with the systematic research of French

## The History of Aphallia and the Intersexual Challenge to Sex/Gender

naturalist Isidore Geoffroy Saint-Hilaire (1832–6). The scientific study of hermaphroditism intensified in the nineteenth century as physicians debated how to distinguish “true” hermaphroditism from “pseudohermaphroditism.” A neo-Aristotelian thinking underlay the belief at the time that most individuals with ambiguous genitalia were really pseudohermaphrodites since they had a “true” sex (either female or male), whereas few people had *both* male and female gonads qualifying them to be “true hermaphrodites.” The challenge was how to determine the presumed true sex.

Given the high risk of exploratory surgery until the twentieth century, physicians relied on anatomical features and stereotypes of gendered behavior and psychology. Alice Dreger examines how Victorian doctors struggled to determine the sex of ambiguously sexed adults based on genital examinations, but even more importantly, the relative masculinity or femininity of their temperament and behavior.<sup>12</sup> The sex of the patient’s erotic partners was often a dominant criterion since doctors sought to prevent the “sexual perversion” of a same-sex alliance.

Eighteenth-century advances in microscopy had allowed for a new perspective on the construction of living organisms. A novel field of biomedical research, histology (microscopic structure of tissue), arose in the early nineteenth century with the work of Marie François Xavier Bichat. Researchers began to understand gonadal histology and were able to visually distinguish testicular from ovarian tissue. By the late nineteenth century this became the gold standard for the sexing of genitally ambiguous individuals.<sup>13</sup> One microscopic surprise was that very few so-called “hermaphrodites” were found to possess *both* testicular and ovarian cells in their gonads. The few that did were designated “true hermaphrodites” whatever their external appearance. Most other ambiguously sexed individuals were found to have either ovarian or testicular tissue; therefore, these patients were designated as “pseudohermaphrodites.” Thus, individuals with ovarian tissue, no matter how masculine their external appearance, were designated female pseudohermaphrodites. On the other hand, individuals with testicular tissue and feminine genitalia were labeled male pseudohermaphrodites. However, given the high risks of exploratory surgery until the twentieth century, histological sexing was generally only practical at autopsy. A neo-Aristotelian, binary thinking pervades this histological approach: that mammals are either male or female and the rest of anatomy – aside from the gonads – is superfluous as far as the essential “true sex” is concerned.

Advances in microscopy also led to the discovery of startling anatomical similarities between early embryos of animals of different species. For example, pigs, rabbits, and human embryos are nearly identical in very early stages of development. This led to Ernst Haeckel’s theory that “ontogeny is the short and rapid recapitulation of phylogeny.”<sup>14</sup> In other words, an individual human’s embryological development repeats the stages of evolution from a one-celled organism through lower orders of animals and mammals before differentiating into a human. Closely related to this was the discovery of the so-called “bi-sexuality” of vertebrate embryos. In early embryonic stages the genitals and gonads are “indifferent,” i.e., can develop into either testes or ovaries. The embryo is also bisexual in terms of the genital ductal system, i.e, both the Müllerian ducts and the Wolffian ducts are present.<sup>15</sup> It is only by the second month post-conception in humans that there is differentiation into typical male or female anatomy.<sup>16</sup> This notion of primitive bisexuality was central to evolutionary

models of phylogeny and ontogeny. Charles Darwin, for example, had hypothesized in *The Descent of Man* that “some extremely remote progenitor of the whole vertebrate kingdom appears to have been hermaphrodite or androgynous.”<sup>17</sup> These models also strongly influenced Sigmund Freud’s model of psychosexual development from primitive, “polymorphous perversity” to civilized heterosexuality, as if the individual recapitulated the sexual evolution of the species.<sup>18</sup>

The term “intersexual” also appeared in the late nineteenth century; however, it referred to the recently described phenomenon of “psychosexual hermaphroditism” or “sexual inversion” – what would later become synonymous with “homosexuality.” These three terms (intersexual, psychosexual hermaphroditism, and sexual inversion) were used almost interchangeably at the turn of the twentieth century. For example, British sexologist Havelock Ellis wrote of “intersexual love” (referring to same-sex love) in his monograph *Sexual Inversion* (1897) and American psychologist G. Stanley Hall wrote of “intersexual attraction” between adolescent boys.<sup>19</sup> In the same vein, Edward I. P. Stevenson (under the pseudonym Xavier Mayne) first used “intersexes” in 1908 as the title of his book subtitled *A History of Similisexuality as a Problem in Social Life*. His work is a defense of same-sex attraction that relies on biological models of bisexual embryological development to argue that “similisexuality” belonged to a “series of originally intermediary sexes – the so called intersexual theory – rather than mere aberrations, degeneracies, psychic tangents, from the male and female.”<sup>20</sup> The first use of the term “intersexual” to denote diverse forms of anatomical sexual ambiguity or atypicality was in 1917 by Richard Goldschmidt in an article on the endocrinology of hermaphroditism.<sup>21</sup>

In 1905, zoologist Edmund B. Wilson and biologist Nettie Stevens independently proposed that distinct X and Y chromosomes determined sex. Wilson discovered the X chromosome in a butterfly while Stevens identified the Y chromosome in a beetle. Karyotyping (i.e., the visualization and organization of an individual’s chromosomes) could be done in a reliable way in the 1950s. With karyotyping, sexing became predominantly a genetic matter.<sup>22</sup> Most female humans have a pair of X chromosomes, while males have an X and a Y. The popular press, the general public, and some judges generally stop at the sex chromosomes in their understanding of sex determination.<sup>23</sup> As with the gonadal gold standard of sexing, the chromosomal standard betrays neo-Aristotelian foundations: an individual has a “true sex” (despite genital anatomy or gender identity) that is either female (XX) or male (XY).

Among biomedical researchers, however, the genetics of sex continues to be an ever-more complex challenge. The dominant hypothesis throughout much of the twentieth century was that once the bipotential gonads differentiated into a testis, the testes produced the necessary hormones for the remainder of male sex differentiation of the sexual organs. A quarter-century quest for a testis-determining factor on the Y chromosome was finally completed in 1990 with identification of the SRY gene (sex-determining region of the Y chromosome).<sup>24</sup> However, since 1990, testis and ovary determination have proven to be far more complex than a single gene and appear to rely on over a dozen genes on the sex chromosomes as well as the other 22 pairs of chromosomes (designated the “autosomes”).<sup>25</sup>

The clinical management of intersexed newborns has long been a medical dilemma. Historically, as noted above, sex assignment was based on external anatomy. However,

## The History of Aphallia and the Intersexual Challenge to Sex/Gender

the availability of chromosomal testing, hormone assays, and non-invasive imaging (such as ultrasound) offer additional data that both informs and further complicates the determination of sex and the prediction of future gender. Today we understand much about the embryology, endocrinology, and molecular basis of the most common etiologies of intersex conditions, including congenital adrenal hyperplasia (CAH), androgen insensitivity syndrome (AIS), 5-alpha-reductase deficiency, and other genetic disorders of sex steroid synthesis. While such biological understanding helps inform a decision about the sex assignment of an intersexed newborn, it has not guaranteed that this neonatal or subsequent sexing matches the individual's self-determined gender identity in childhood or adulthood. Historically, in certain published cases, erroneous sex assignment has led to tremendous psychological suffering.<sup>26</sup>

The "optimal gender" paradigm for sex determination is based on a model of gender and its plasticity first established at the Johns Hopkins Psychohormonal Research Unit. In the 1950s, Hopkins psychologist John Money developed the conceptual distinction between "sex" (the biological and anatomical attributes of male and female) and "gender" (the psychological and socio-cultural aspects).<sup>27</sup> Furthermore, he made the distinction between "gender identity" (self-identification as male, female, or ambivalent) and "gender role" ("everything that a person says and does, to indicate to others or to the self the degree that one is either male or female or ambivalent; it includes but is not restricted to sexual arousal and response").<sup>28</sup>

Based on ethological research on imprinting and their own experience with hermaphroditic and genitally ambiguous infants, the Hopkins researchers concluded that gender identity was entirely socially malleable during an early period of plasticity. They argued that

the sex of assignment and rearing is consistently and conspicuously a more reliable prognosticator of a hermaphrodite's gender role and orientation than is the chromosomal sex, the gonadal sex, the hormonal sex, the accessory internal reproductive morphology, or the ambiguous morphology of the external genitalia.<sup>29</sup>

Therefore, they argued that infants born with ambiguous genitalia could be surgically "corrected" and then successfully raised as either males or females so long as certain conditions were met:

- 1 gender assignment was done before 18–24 months;
- 2 the parents were not ambivalent in the gender of rearing; and
- 3 the children were not confused by knowledge about their intersexed past.<sup>30</sup>

Relying on results with sex reassignment in transsexuals, Money and his colleagues claimed that hormonal treatment beginning in puberty could complete the development of secondary sex characteristics and give an outwardly normal sex appearance. Money and Ehrhardt illustrated their claims with the case of a "genetic male hermaphrodite" who, after undergoing orchiectomy (surgical removal of the testes) and initiating estrogen therapy even at eleven years of age, still had a good outcome: "The result is a perfectly feminized body, indistinguishable in morphology and appearance from that of a genetic female with her own ovarian puberty – though menses, of course are lacking."<sup>31</sup>

Money and his colleagues originally proposed that the appearance and appropriate functioning of the genitals was a dominant factor in assigning sex: “a great deal of emphasis should be placed on the morphology of the external genitals and the ease with which these can be surgically reconstructed to be consistent with the assigned sex.”<sup>32</sup> Effectively, this meant that doctors recommended that intersexed infants with phalluses deemed too small (i.e., less than 2.5 cm) be reassigned as female because it was felt that it would be an intolerable psychic burden to be a male with a so-called micropenis. On the other hand, doctors recommended that an enlarged clitoris (i.e., larger than 1 cm), be “reduced”; although frequently this amounted to clitoridectomy.<sup>33</sup> Again, this was because, presumably, it would be psychologically damaging for a girl to have a phallus-like clitoris. The Hopkins team argued that, “Clitoral amputation in patients living as girls does not, so far as our evidence goes, destroy erotic sensitivity and responsiveness, provided the vagina is well developed.”<sup>34</sup> Unfortunately, there was little long-term follow-up to establish the accuracy of this assertion. In all cases, it was essential that a definite sex assignment be made early (preferably within the first few days, and at the latest by two to three years of age) and that the parents not be left with any uncertainty about what gender to raise the child. The child should also be protected from any knowledge about his or her intersex past to prevent gender confusion. The new standard of care that gradually developed thanks to Money’s work in the 1950s and still dominates has been termed the “optimal gender” policy.<sup>35</sup> Under this approach sex assignment is based on a variety of factors: reproductive potential (if possible), good sexual functioning, minimizing medical procedures, convincing gender appearance, stable gender identity, and psychosocial well-being.

The linchpin of Money’s theories was the now infamous case of John/Joan (actually named Bruce/Brenda Reimer). Bruce was a non-intersexed boy, one of a pair of monozygotic (identical) twins. Due to penile phymosis (constriction of the foreskin), Bruce underwent circumcision at 7 months. Because of surgical instrument malfunctioning, his penis was severely burned and eventually sloughed off. Upon Money’s recommendation, the parents agreed to reassign their son as a girl. Through multiple interventions beginning at 17 months and continuing through puberty, the child underwent orchiectomy, partial vaginoplasty (surgical vaginal construction), and estrogen therapy. When Money and Ehrhardt discussed the case in *Man and Woman, Boy and Girl* (1972), they reported that the child (then age 7) had been successfully reared as a girl.<sup>36</sup> Money later glowingly declared that at age 9, Brenda’s “behavior is so normally that of an active little girl, and so clearly different by contrast from the boyish ways of her twin brother, that it offers nothing to stimulate one’s conjecture.”<sup>37</sup> Money last saw Brenda in 1979 and then the family was reportedly “lost to follow-up.” Keith Sigmundson and Milton Diamond tracked down “Brenda” in the early 1990s, and reported that Money and Ehrhardt’s sanguine assessment of the case was thoroughly unwarranted.<sup>38</sup> Brenda had rebelled against her gender assignment, had a terrible time fitting in at school, and was frequently suicidal. At age 14 when she refused further hormonal and surgical interventions she was finally informed about her true entire medical history. Brenda decided to become “David” and underwent androgen therapy, a mastectomy, and phalloplasty. He reportedly transitioned quickly into a male role and married at 25, adopting his wife’s children.<sup>39</sup> Tragically, David

## The History of Aphallia and the Intersexual Challenge to Sex/Gender

Reimer committed suicide on May 4, 2004 after having lost his job and separated from his wife. He had also been grieving the death, two years previously, of his twin brother.<sup>40</sup>

It is precisely because of cases like this and others where intersexed adults were angered by disfiguring surgeries with lingering pain that the Intersex Society of North America (ISNA) was launched in 1993 by Cheryl Chase. ISNA members also deplored the paternalizing misinformation sponsored by their families and doctors. Thanks to the internet, ISNA and other intersex organizations serve as international information sources for intersexed individuals, their families, and loved ones. ISNA is actively and quite successfully engaged in lobbying physicians and medical organizations to change the interventionist model of treating intersexuality. Like Diamond and Sigmundson, ISNA favors surgical interventions only in life-threatening situations (which are rare) and deferring elective surgeries until the child can decide about them.<sup>41</sup> Growing numbers of physicians are adopting this more conservative approach to intersex surgery until research studies can substantiate the functional and psychological benefits of early genital reconstructive surgery.<sup>42</sup>

## The Medical History of Penile Agenesis

While 1 in 1000 to 2000 neonates are born with anatomically ambiguous genitalia, penile agenesis or “aphallia,” is an extremely rare phenomenon.<sup>43</sup> A recent review article on aphallia noted approximately 60 cases reported in the entire medical literature.<sup>44</sup> A more restrictive review of the literature from 1966 to 2004 uncovered 33 cases.<sup>45</sup> *Campbell's Urology* textbook gives an incidence of one in 30 million males.<sup>46</sup>

The first published case I have discovered dates to 1701 and was recorded by the French surgeon Barthelemy Saviard (1656–1702). Under the heading of a “child who had no rod,” Saviard described a newborn child born with a small, erectile prominence – like the “rump of a hen” – in place of a penis. This structure had a urinary meatus (urinary outlet) and, two months after birth, the phallus had visibly grown. Saviard identified the child as a male without hesitation or even any consideration of hermaphroditism. Instead Saviard offered a possible etiology that was in keeping with centuries of medical theorizing about the power of the imagination in the generation of “monsters.”<sup>47</sup> During the pregnancy, the mother had had a tremendous craving for chicken but had not been able to satisfy this appetite. Saviard explained that her preoccupation with chickens had caused the penile deformity in the shape of a hen's rump. While this seems like a fanciful notion today, it nevertheless attests to the power attributed by Renaissance doctors to psychological forces over bodily physiology. Also of particular importance in this case is that Saviard clearly presents this as a male infant with a penile birth defect not as a hermaphrodite. At the time, surgical sex reassignment was inconceivable and there was no question of assigning this infant a female sex.

The embryogenesis of the urogenital system was elucidated beginning in the nineteenth century, and now is better understood in terms of its anatomical development and endocrinological basis.<sup>48</sup> As noted earlier, a striking discovery was that both male and female genitals develop from the same undifferentiated primordia. The

genital tubercle, which begins to form in the third week in both males and females, begins to develop in the tenth week into a phallus under the influence of 5-alpha-dihydrotestosterone. In the presence of estrogen and low levels of androgens (or in androgen insensitivity syndrome), the genital tubercle will take on a clitoral configuration. Agenesis of the penis is a result of complete or partial failure in phallic development of the genital tubercle and failure in caudal migration of the urogenital sinus. Therefore, aphallia is accompanied by an anomaly of the urethral meatus (urinary outlet): urethral atresia (narrowing or absence) or placement anterior to the anus or within the rectum.<sup>49</sup> In other words, there may be no exit for urine or it may exit anomalously at the perineum or within the rectum.

The surgical treatment of penile agenesis has undergone a dramatic shift, which is evident from examining sequential editions of *Campbell's Urology*, one of the leading American textbooks in the field. In the third edition, the chapter on "Anomalies of the genital tract," written by Meredith Campbell herself, concludes that "there is no satisfactory treatment."<sup>50</sup> She cites surgeon Harold Gilles's attempts in the 1940s at phalloplasty for war injuries, but notes that the most which can be hoped for is an acceptable cosmetic result but not a functional penis.<sup>51</sup> In the fifth edition of the textbook, however, Drs Duckett and Snow conclude the brief section on penile agenesis with an unwavering recommendation: "Female gender assignment is recommended in the newborn period and elective orchiectomy should be performed prior to puberty."<sup>52</sup> Skoog and Belman, in their extensive review of aphallia, support this policy of early sex reassignment by referring to the research of John Money (discussed above) and an article by Hugh Hampton Young, Robert Stoller, and colleagues.<sup>53</sup>

Psychoanalyst Robert Stoller, a professor of psychiatry at the University of California, Los Angeles Neuropsychiatric Institute, was a pioneer in the theorization of gender identity, transsexualism, and sadomasochism from the 1960s until his untimely death in 1991. In 1964, he described a case of a "hermaphroditic" adolescent who had been brought up as a girl, but had always been a tomboy. When her voice deepened at age 14 she underwent a medical examination that demonstrated 46 XY karyotype (the usual male chromosome pattern), clitoral penis, hypospadias (misplaced urinary opening), bilateral cryptorchidism (undescended testes), and bifid scrotum (split in two, giving a labial appearance). Reportedly, the girl took this news without surprise and immediately shifted to a male gender role. School performance, interactions with parents, and overall psychological adjustment improved dramatically. Stoller coined the notion of a male "core gender identity," which in this case explained the patient's effortless gender shift. He argued that this unalterable sense of gender identity was established prior to the Oedipal stage, contrary to classical analytic teachings.<sup>54</sup> This case suggested that male core gender identity could form in the absence of a penis and despite unequivocal parental rearing as female. Stoller, therefore, seriously entertained the possibility of "a congenital, perhaps inherited biological force" in the production of gender identity.<sup>55</sup>

Later the same year, Stoller published the case of an adult "hermaphrodite" in psychotherapy.<sup>56</sup> The patient had initially presented herself to Stoller as a butch lesbian in her mid-forties, who had always been aware of having been born with an "enlarged clitoris" that had never been surgically altered. Stoller reports that thanks to psychotherapy she rediscovered her childhood hermaphroditic identity and finally agreed to

## The History of Aphallia and the Intersexual Challenge to Sex/Gender

a thorough medical workup that showed she possessed testicles. The patient transitioned gender role to male, left the lesbian community, changed the sex on his birth certificate, and married. Stoller's conclusion from the case was that core gender identity is not limited to male and female, but that a "hermaphroditic identity" is possible when parents are uncertain about their child's sex and the child becomes aware of his or her genital uniqueness. Furthermore, Stoller proposed that only individuals with this sort of childhood hermaphroditic identity could make an adequate adolescent or adult adjustment to sex reassignment. On the other hand, Stoller argued that intersexed individuals with an unwavering male or female core gender identity would be emotionally devastated by the information that they are biologically of the opposite sex, and would never be able to transition genders successfully.

Stoller described two cases of penile agenesis in children aged 4 and 15 in 1965. Both children had clear and unambiguous male gender identity. The 4-year-old, who had a perineal urethrostomy (surgically-created urinary opening) and no genital surgery, was vehemently opposed to the idea of sex reassignment to female. The 15-year-old had undergone multiple surgical procedures resulting in a severely deformed phallus that Stoller described as a "monstrosity of unearthly appearance." Stoller did not recommend sex reassignment of these children, but instead saw them as confirming his earlier claim about the fixity of "core gender identity." Furthermore, these cases supported his argument that the anatomic penis was not essential to the "sense of maleness." Stoller, however, was already greatly impressed by the early work of Money and the Hampsons, which he viewed as "most compatible with present-day psychodynamic opinion" and therefore reiterated their claim that "gender role is determined by postnatal psychological forces, regardless of anatomy."<sup>57</sup> In his two cases of aphallic children, Stoller argued that the parents' gendered rearing of the infants had produced a male identity.

In 1968, Stoller's belief in a biological determinant of gender identity changed dramatically. He published an update on "Agnes," the case of a female-identified teenaged boy who initially claimed to have spontaneously feminized at puberty. Although a thorough medical workup failed to establish a diagnosis, the boy was hypothesized to have a hermaphroditic condition that would explain the female gender identity and bodily feminization. At the patient's request, she underwent female sex reassignment at age 20.<sup>58</sup> Several years later Agnes reemerged and admitted to Stoller that she had deceived her medical and psychiatric teams. In fact, she had been secretly taking her mother's estrogen pills since puberty. In 1968, Stoller revised Agnes' diagnosis from hermaphroditism to male transsexualism and now rejected the role of a "biological force" in shaping core gender identity. Instead, he emphasized a particular constellation of pathological parent-child dynamics in effeminate boys: their mothers had been tomboyish, the father was passive and effeminate, both parents were permissive about the child's effeminacy, and the mothers had excessive contact with the boy.<sup>59</sup>

This significant shift in his thinking is evident in a 1971 article with pediatric urologist Hugh Hampton Young, a leader in the surgical treatment of intersexes. By 1971, Stoller supported female sex reassignment of neonates with penile agenesis. Discussing four cases of aphallia, Young and Stoller recommended that a team including a urologist, endocrinologist, and psychiatrist make a diagnosis within a few days of birth and that the female sex assignment be presented "before the parents are aware

of the problem.”<sup>60</sup> They believed that orchiectomy could be deferred if there was a good likelihood of close longitudinal follow-up. Relying on their experience with Agnes, they believed that aphallic children could undergo later genital feminization and estrogen therapy resulting in completely normal female identity, appearance, and sexual functioning.<sup>61</sup> They concluded, echoing Money’s team, that gender identity is established by the parents and that female rearing would lead to easier and better psychological development and peer relations.

Money and Ehrhardt reiterated this recommendation in cases of agenesis or traumatic loss of the penis because, “With an appropriately timed program of surgical and hormonal correction, the baby’s core gender identity will then differentiate as female.”<sup>62</sup> Kessler and McLaughlin at the University of California, San Diego, recommended orchiectomy within the first few years of life and vaginoplasty in adolescence, confident that this would lead to successful female identity and sexual functioning.<sup>63</sup> In 1987, Oesch, Pinter, and Ransley reported on six aphallia cases, recommending early female sex assignment and gonadectomy in the first few days of life.<sup>64</sup> Skoog and Belman recommended that orchiectomy, labial reconstruction, and urethral transposition be done in the neonatal period.<sup>65</sup>

Among these publications increasingly pushing for female sex assignment of individuals with penile agenesis, there is one highly unusual case. In 1973, Rosenblum and Turner described a 45-year-old black man from rural South Carolina with no prior medical care, who was referred to urologists because of congenital absence of the penis. He was married and reported satisfactory marital relations. He claimed he could have intercourse and ejaculate by stimulating the dorsal portion of his scrotum and pubic area. Psychological testing found that he had “adjusted to his situation in a reasonable fashion.”<sup>66</sup> He refused to have any surgical “corrective therapy” and did not return for follow-up. Ironically, this person’s poverty and rural residence probably preserved him from neonatal sex-reassignment and forced him to adapt (apparently successfully) to his genital anomaly.

Aside from Stoller’s case descriptions from the 1960s, there are few accounts of psychotherapy with intersexed individuals, particularly children. This is probably because of the conspiracy of secrecy around their past.<sup>67</sup> Stoller, Money, and others who promoted the “optimal sex” paradigm counseled against informing intersex patients about their intersex diagnosis and past genital surgeries for fear that this information could lead to gender confusion. Doctors and families in the know were to maintain a paternalistic veil of secrecy. The intersex individuals were, nevertheless, likely to realize that something was unusual about their genital anatomy or sexual function, and undoubtedly were likely to be suspicious about their frequent medical visits, surgical revisions, and maintenance hormone therapy. However, the atmosphere of secrecy would have encouraged them not to inquire about their medical condition, or worse, to be profoundly ashamed of it. It is quite likely that many intersex individuals have been in therapy over the past four decades, but never mentioned it to a therapist because they were unaware of or were too embarrassed to bring up their intersex condition. They may also fear that a clinician will become overly fascinated with or distracted by the intersex history and fail to provide the services the patient desires.<sup>68</sup> Equally worrisome are cases where a therapist who was privy to the patient’s intersex history felt compelled to maintain the secret. How can a thera-

pist work successfully with someone while withholding critical information from the patient? Intersex management teams may also avoid referring their patients for psychotherapy precisely out of fear of having them learn about their past.

### Intersex and Gender Identity

Let me return, in concluding, to my patient Mario. In clinical settings I have discussed in detail the course of his psychotherapy and the challenging psychosexual development he is undergoing as a pubescent boy with surgically constructed female external genitalia. Rather than focus on his case (since few of my readers in this context are psychotherapists), I want to concentrate on the broader theoretical issues of how the history of intersex treatment and current intersex politics affects children with aphallia and other intersex conditions. First, it is essential to know the medical history of hermaphroditism to understand the psychological and biomedical logic that dictated Mario's past treatment. Although the management of intersexes is undergoing significant reconsideration now, partly under pressure from intersex advocacy groups (Chase 1999),<sup>69</sup> many intersexed children are still assigned a sex and surgically "corrected" based on the paradigm shaped by John Money, Robert Stoller, and other researchers in the 1960s. Their central tenet of the complete psychosocial malleability of gender is coming under increased scrutiny as researchers examine sexual dimorphism in the brain.<sup>70</sup> Diamond hypothesizes that gender identity is strongly determined *in utero* by the influence of sex steroid levels on the developing brain.<sup>71</sup> Even John Money no longer claims that rearing *exclusively* determines gender identity.<sup>72</sup> In 2005, Heino Meyer-Bahlburg reviewed penile agenesis cases in the medical literature and their gender outcomes. Remarkably, he found that few case descriptions even systematically assessed gender identity or dysphoria. Of those that did, 16 patients had been assigned female and 17 assigned male. None of those assigned male expressed gender dysphoria or had switched to living as females, whereas 4 out of 16 patients assigned female expressed gender dysphoria or had switched to a male gender role. Given the relative novelty of the "optimal gender" paradigm, only a minority of these patients had even reached 18 years of age and therefore had further psychological development ahead of them when they might become dissatisfied with their assigned gender. Meyer-Bahlburg, a long-time Money collaborator, cautiously concluded that while social factors in gender assignment play a role, female assignment of children with penile agenesis carries a risk of later gender dysphoria and gender change.<sup>73</sup> Zucker was more decisive in concluding that female sex assignment is *not* appropriate treatment in cases of penile agenesis (such as Mario's), traumatic loss of the penis, or micropenis since current accumulated evidence indicates that these infants later identify as male in adulthood.<sup>74</sup>

Stoller's original concept of core gender identity re-emerges as accurate and, in Mario's case, his mother recalls that he displayed strong male typical behavior since infancy and he always said he felt like a boy. As Stoller first suggested, a firm male core gender identity can develop in early childhood without a penis or, in Mario's case, despite the presence of surgically constructed labioform genitals. Furthermore, Mario's male gender developed despite attempts at enforced female socialization in

the first three years of life. Contrary to Stoller's model, it does not currently appear that this knowledge led Mario to an "intersex identity." He has a strong male core gender identity while being fully cognizant of his anatomical difference compared to other boys.<sup>75</sup>

Preschool children certainly manifest a broad spectrum of gendered behaviors. Gender role is heavily policed by parents as well as peers and conventional gender roles become more polarized as children age. While gender variant children (so called "sissy boys" and tomboy girls) challenge the dichotomized gender roles, it is only rarely that even these children are uncertain about their sex. Many adult gays and lesbians also recall feeling vaguely "different" in terms of their gender as children, without having felt confused about their sex.<sup>76</sup> Young children, at least in a Euro-American context, do not seem capable of sustaining such a novel and culturally atypical third gender identity as being a hermaphrodite or intersex. Stoller's formulation of the "hermaphroditic identity of hermaphrodites," however, may be a retrospective construction by adults that is important, if not essential, to adult intersexuals' gender identity transitioning.<sup>77</sup> Some intersexual activists, such as Howard Devore, have declared their own intersex gender identity as both male and female or a third gender between male and female.<sup>78</sup> However, the mainstream intersex support groups centered around particular diagnoses (such as androgen insensitivity syndrome, hypospadias, or congenital adrenal hyperplasia) have intensely debated if not completely rejected the intersex label because the affected individuals feel their gender identity is either male or female and they do not want to be perceived as gender intermediates. Preves also notes that her informants, recruited from intersex support groups, although developing an identity as intersexed people (that is, they acknowledged to themselves and publicly that they had an intersex condition), nonetheless maintained a binarized view of gender and "went to great lengths to uphold the sex of assignment they were given."<sup>79</sup>

Contrary to what some members of these support groups believe, the ISNA does not promote intersex as a third gender or inter-gender identity. The misperception probably arose from ISNA's in-your-face activism in its first years, when it used the attention grabbing phrase "Hermaphrodites with Attitude." The gender radicalness that intersex support groups reject has instead been exploited by some feminist theorists who have seen in the intersex condition a tool for demolishing cultural and medical constructions of sex and gender. Suzanne Kessler, for example, utopically concludes her monograph declaring: "We must use whatever means we have to give up on gender. The problems of intersexuality will vanish and we will in this way compensate intersexuals for all the lessons they have provided."<sup>80</sup> Early in the intersex battles, Anne Fausto-Sterling had argued that intersexes demonstrated the spectrum of human sexuality and proposed three terms for intersex sex identities.<sup>81</sup> After dialogue with Cheryl Chase, Fausto-Sterling later retracted that essay as a "tongue in cheek" provocation.<sup>82</sup>

Judith Butler also made use of the intersex phenomenon in a theoretically laden essay on the "desubjugation of the subject within the politics of truth."<sup>83</sup> Her article, unfortunately, simplified intersex and misrepresented most of her sources. She claimed, erroneously, that Cheryl Chase argues that "there is no reason to make a sex assignment at all."<sup>84</sup> Butler also unfairly portrayed Milton Diamond as a simplis-

## The History of Aphallia and the Intersexual Challenge to Sex/Gender

tic Y chromosome determinist who argues that any infant with a Y chromosome be assigned or reassigned male.<sup>85</sup> Most ironically for an article entitled “Doing Justice to Someone,” Butler persistently referred to David Reimer as “John/Joan” for the sake of her discourse theory argument that “he is the human in its anonymity . . . [H]e is the anonymous – and critical – condition of the human as it speaks itself at the limits of what we think we know.” Only in a footnote did she note that “John/Joan no longer operates with a pseudonym,” yet she never names David Reimer.<sup>86</sup> In a subsequent revision of the essay in *Undoing Gender* (2004), Butler has replaced Reimer’s name throughout without, however, altering her conclusion on anonymity or correcting misrepresentations of the biomedical literature. She still sees in the intersex condition an opportunity for destabilizing biological notions of sex and gender.<sup>87</sup>

ISNA, on the other hand, has struggled to shift the focus of intersex politics from sex/gender theory battles to practical clinical concerns. Its home webpage in 2004 made its position on gender unmistakably clear: “WARNING! *Do not* claim that ISNA is or ever has been in favor of three sexes, or no sexes, or eliminating gender.”<sup>88</sup> Cases of sex reassignment, such as Mario’s or David Reimer’s, are quite rare and not the central focus of ISNA, despite the huge media and academic attention they attract. ISNA’s mission is to reduce the stigma and secrecy surrounding intersex conditions and eliminate unnecessary intersex pediatric surgeries. The vast majority of these surgeries are done, not to change an infant’s sex, but to make the genitals appear more “normal,” that is, more sex-typical. Kessler accurately criticized the sex bias underlying this surgical normalization: girls should not have a significant clitoris and boys should have a large penis for future vaginal penetration.<sup>89</sup>

It is as yet not clear how gender identity is determined. There may be genetic factors, *in utero* hormonal effects, as well as rearing effects. All three probably play a role. But contrary to Money’s original hypothesis, gender identity most likely is not entirely malleable through rearing and genital surgery. Given that in the vast majority of *all* individuals gender identity is quite robust by age 2 or 3 (even if discordant with genital sex) there seems little logic to the policy of secrecy surrounding intersex conditions. A little girl informed of her CAH diagnosis is unlikely to have a gender crisis and decide she is really a boy. The more pressing issue is coping with the ongoing medical attention and medications she needs to stay healthy for the rest of her life. Being kept ignorant about her CAH diagnosis is not likely to help her attend to her own health care. The secrecy surrounding an intersex diagnosis and any early corrective surgery, therefore, seems misguided and likely to stimulate a distrust of medical care and reinforce the child’s feeling that genitals and gender status are unmentionable and shameful – a common experience for intersexed individuals.<sup>90</sup>

Testimonials from the intersexed members of the ISNA suggest that those who had surgical genital interventions always had a feeling that something was wrong with them.<sup>91</sup> They had to make sense of repeated doctor visits, genital examinations, unusual genital appearance, scarring, and sensory deficits. The frequent genital examinations alone can be experienced as sexual molestation.<sup>92</sup> In other words, the paternalistic policy of secrecy, while it may have been well-intentioned, does not succeed in letting these children feel “normal.” Medical reassurances that an intersexed child will be a “normal” adolescent or adult after surgical interventions, may assuage parents, but are not founded on long-term follow-up studies.

There are no simple solutions to the management of infants with atypical or ambiguous genitalia. Even without surgical interventions, periodic medical evaluations of genital development, gonadal anatomy and function, or urinary flow are likely to be traumatic for children.<sup>93</sup> Wilson and Reiner have suggested a tactic of sex assignment based on historical experience and suspected prenatal neurohormonal exposure, with only truly essential surgical intervention.<sup>94</sup> They recommend that surgical genitoplasty be deferred until the patient can participate in a discussion about it. Pubertal hormone therapy should also take into account the patient's self-defined gender identity.

The clinical realities faced by intersexed individuals and their families are enormously challenging. Intersexed conditions are a diverse collection of anatomical and physiological atypicalities that in some cases require life-saving corrective surgery and in some cases require life-long endocrinological care. While the historical construction of sex, gender, and hermaphroditism certainly inform the current "optimal gender" paradigm of treatment, deconstructing these will not make intersexuality disappear any more than it will erase the categories of sex and gender. No amount of theorizing about intersex or its cultural impact on gender theory will eliminate the physical pain, infertility, endocrinological disorders, and emotional distress that burden many people with intersex conditions. However, the critical study of intersex can help spur better scientific research and clinical management, as well as spare intersexed individuals unnecessary surgeries, clinical disinformation, and societal marginalization.

## Acknowledgments

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## Notes

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## The History of Aphallia and the Intersexual Challenge to Sex/Gender

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The paired Wolffian (or mesonephric) ducts usually develop into the male reproductive ducts connecting the testes to the exterior: epididymis, vas deferens, seminal vesicle, and central zone of the prostate. The peripheral and transitional zones of the prostate develop from the urogenital sinus. In females the Wolffian duct usually degenerates, leaving behind a remnant: the Gartner duct.
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## The History of Aphallia and the Intersexual Challenge to Sex/Gender

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## The History of Aphallia and the Intersexual Challenge to Sex/Gender

or in a third gender?” The answer still is: “No, and for the record, we’ve never advocated this.” [www.isna.org/faq/third-gender](http://www.isna.org/faq/third-gender). Accessed January 9, 2007.

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